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# Social participation of adult patients with spinal muscular atrophy: frequency, restrictions, satisfaction and correlates.

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**ABSTRACT**

# Social participation of adult patients with spinal muscular atrophy: frequencies, restrictions, satisfaction and correlating factors.

**Introduction** We assessed social participation in 62 adults with spinal muscular atrophy (SMA) types 1c-4.

**Methods** Outcome measure: Utrecht Scale of Evaluation Rehabilitation-Participation (USER-P) with Frequency, Restrictions and Satisfaction scores. Hierarchical regression analysis.

**Results** Early (type 1,2,3a) and late onset (type 3b,4) SMA patients reported similar frequency and satisfaction scores. 'Age', 'motor skills', 'pain' and 'feelings of depression' correlated with frequency; 'motor skills' and 'feelings of depression' correlated with restrictions and 'level of education', 'fatigue' and 'feelings of depression' correlated with satisfaction. Motor skills and feelings of depression explained 33% of variance in *frequency* of participation. Motor skills explained 26% of variance of *restrictions* in participation. Fatigue and feelings of depression explained 50% of variance in *satisfaction* with participation.

**Discussion** Motor skills, feelings of depression and fatigue are correlates of participation in daily life. This knowledge can be used to optimize care for SMA patients.

Key words: social participation, satisfaction, social engagement, activities of daily living, SMA, adults

## INTRODUCTION

Hereditary proximal spinal muscular atrophy (SMA) is an important genetic cause of disability in childhood and adult life.[1] The antisense oligonucleotide nusinersen has been approved and has shown efficacy in altering the natural history of the disease.[2] Other gene therapy trials are ongoing. However, there is currently no cure for SMA.[3,4,5]

The variety and severity of impairments and disabilities that accompany SMA have been described extensively.[6-10] The consensus guidelines for supportive care outlines management for the most common medical complications (e.g. pulmonary problems, scoliosis) that occur primarily in childhood, but focus strongly on symptom management.[11] Irrespective of SMA type, all patients will encounter moderate to severe disability in life. There is, however, a striking lack of literature on how adult patients participate in daily life activities and how multidisciplinary care can optimize their participation. The few existing studies on participation among adult patients with a broad range of neuromuscular disorders have focused mainly on work, and the results of SMA patients were not reported separately.[12-15] In another qualitative study, SMA patients rated their quality of life as 'fine', but experienced the serious impact of progressive functional limitations on their daily activities.[16] Patients emphasized their need to live a normal life and fully participate in social activities, including an active family role, work and maintaining optimism.[17] Increasing our knowledge of participation among adult patients with SMA and factors associated with participation may help to optimize supportive care.

Therefore, the first objective of this study was to describe the frequency of, and perceived restrictions in, participation and the related satisfaction levels of adult patients with SMA. The second objective was to determine whether selected subjective complaints (pain, fatigue, anxiety, feelings of depression) and coping style are associated with participation in adult patients with SMA, adjusting for demographic factors and disease severity. These possible

correlates were selected based on studies showing associations between pain and fatigue, emotional distress, mood and coping strategies with participation and quality of life among patients with neuromuscular diseases.[18-21]

## METHODS

### Subjects and Procedures

Adult patients with genetically confirmed SMA, regardless of type of SMA, aged 18 years or older at inclusion, were recruited for this study between September 2010 and December 2012. Patients were informed about the study and recruited through the Dutch patient organization for neuromuscular diseases (www.spierziekten.nl), through patient communities on the internet, through pediatricians, (pediatric) neurologists, rehabilitation physicians and the four Dutch Centers for Chronic Respiratory Ventilation. The only exclusion criterion was the inability to read Dutch. All patients were retrieved from the Dutch SMA database.[22] For the purpose of this study, all patients were seen at the outpatient clinic of the Department of Neurology and Neurosurgery at the University Medical Center Utrecht for a structured interview and neurological examination. They were asked to complete questionnaires.
This study was registered in the Dutch registry for clinical trials (study no. NL29692.041.09/29692). The Medical Ethics Committee of the University Medical Center Utrecht approved the research protocol. All patients gave informed consent prior to inclusion.

### Measures

The SMA classification system was used to define SMA types 1-4 [6,23,24]
Participation was measured using the Utrecht Scale for Evaluation Rehabilitation-Participation (USER-P), a self-report instrument with 32 items. Validity and reproducibility of the USER-P scale are good.[25-29] Sum scores are calculated for the Frequency, Restrictions and

Satisfaction scales, and each sum score is converted to a score on a scale ranging from 0 to 100. Higher scores indicate more favourable levels of participation (higher frequency of activities, fewer restrictions, greater satisfaction). An example of a question for the Frequency scale is: "How many hours do you spend on household activities?"

An example of a question for the Restrictions scale is: "Does your illness or condition currently limit your daily life concerning outdoor mobility?". An example of a question for the Satisfaction scale is: "How satisfied are you with your current daily life concerning 'going out' (eating out, visiting a cafe, the cinema, a concert, alone or with others)."

**Correlates**

We used a standardized questionnaire to document disease characteristics (age at diagnosis, age at loss of ambulation) and demographic characteristics (sex, age, relationship status, job status, level of education, whether living independently).

To document motor skills we used the Expanded Hammersmith Functional Motor Scale (HFMSE), a validated test consisting of 33 items to assess motor skills of patients with SMA.[30,31] The maximum score is 66 points. Higher scores indicate better motor skills.

To assess pain and fatigue we used the sub-domain scores 'pain' (two items) and 'vitality' (four items) of the Short Form 36-item Health Survey (SF-36).[32,33] The vitality score is a valid instrument to measure fatigue, as shown by a high correlation between the SF-36 vitality score and scores on the Fatigue Symptom Inventory.[34] Higher scores in the domain 'pain' indicate that patients experience less pain; higher scores in the domain 'vitality' indicate that patients experience less fatigue and feel more vital.

Feelings of depression and anxiety were assessed using the Hospital Anxiety and Depression Scale (HADS). [35-37] Seven items assess feelings of anxiety (HADS-A) and seven items assess feelings of depression (HADS-D), with a maximum of 21 points for the HADS-A and HADS-

D scales. A score of 8 or more on the HADS-A or HADS-D is an indication of anxiety or depressed mood.

Coping strategies were assessed using the short version of the Coping Inventory for Stressful Situations (CISS-21).[38,39] The CISS-21 has 21 items divided into three categories: problem-targeted strategies (CISS-P), emotion-focused strategies (CISS-E) and avoidance strategies (CISS-A). Response options range from 'not at all'(1) to 'very applicable'(5). A higher score indicates a preference to use this particular coping strategy.

**Statistical analyses**

Descriptive statistics were used to describe characteristics of the study population and participation scores. Comparisons between all SMA types were not performed because of the small sample size. Since differences in quality of life were found between patients with a relatively early onset (i.e. SMA types 1-3a) and those with onset later in life (i.e. SMA types 3b-4),[40] we dichotomized the variable SMA types to early onset SMA (types 1-3a) versus late onset SMA (types 3-4) to compare outcomes between these two subgroups.

In addition to the total scores on the USER-P, individual items of the Restrictions and Satisfaction subscales were dichotomized to quantify the presence of restrictions in and dissatisfaction with specific aspects of participation. The options "with difficulty", "with assistance" and "not possible" on the restrictions subscale were defined as "restrictions". The option "without difficulty" was defined as "no restrictions". The answer options "satisfied" and "very satisfied" on the satisfaction subscale were defined as "satisfaction". The answer options "very dissatisfied", "dissatisfied" and "neutral" were defined as "dissatisfaction". The answer option 'not applicable' was defined as a missing variable.

To detect differences in determinants of participation between early and late-onset SMA patients, we used the independent samples Mann Whitney U test. Spearman correlations were

computed to determine relationships between potential determinants and participation of the total sample. Using Cohen's rule of thumb, a correlation of 0.10 was considered 'small', of 0.30 'medium' and of 0.50 'large'.[41]

Determinants that showed a p-value < 0.1 in the bivariate correlation analysis were entered into a hierarchical linear regression model (Multi-step enter method). Variables were always entered in the same order: step 1: disease severity variables and demographics; step 2: subjective complaints. Residual analyses were performed and multi-collinearity was tested to search for violations of necessary assumptions in multiple regression.[42] SPSS version 24 for Windows was used for analysis.

## RESULTS

Sixty-two of 80 (78%) invited patients participated. Descriptive statistics are displayed in Table 1. We included four patients with SMA type 1c and onset before 6 months of age who never learned to sit independently but survived into adulthood. Median HADS-A and HADS-D scores were low (Table 2). Patients with early onset SMA had significantly lower motor skills (HFMSE-scores) than those with late onset SMA. Significantly fewer patients with early onset SMA had a partner than patients with late onset SMA. Late onset patients reported more pain and more fatigue than early onset patients.

*Frequency (level) of participation*: Median participation scores are displayed in Table 3. There were no significant differences between patients with early and late onset SMA in the frequency of participation (Table 3). SMA patients spent most hours on unpaid work and household activities (Table 4). Only 39% of them had paid work and only 16% had a full-time job (36 hours a week or more). A detailed description of all participation activities performed by all patients is given in Table 4.

*Restrictions and satisfaction*: Patients with early onset SMA experienced significantly more participation restrictions compared to patients with late onset SMA (Table 3). Table 5 shows that 16-97 % of patients felt restricted in daily activities. They felt most restricted in work/education, household chores, mobility outdoors (e.g. traveling by car, public transport or bike), going out (e.g. to a movie, pub, church, or shopping), physical exercise and visits to family and friends. There were no significant differences between patients with early and late onset SMA in satisfaction with participation (Table 3). Table 5 shows that 8-58 % of patients were dissatisfied about daily activities. They were most dissatisfied about performing household chores and physical exercise.

*Correlates of participation* (Table 6). Bi-variable analysis showed a medium correlation between lower frequency of participation and older age, reduced motor skills and pain. We found a medium correlation between lower frequency of participation and more feelings of depression. Participation restrictions were largely correlated with reduced motor skills. There was a medium correlation between satisfaction with participation and level of education, , fatigue and a large correlation with fewer feelings of depression.

Table 6 summarizes results of the multivariable analysis. 33% of the variance in frequency of participation was explained by motor skills in combination with feelings of feelings of depression; 26% of the variance in restrictions in participation was explained by motor skills and 50 % of the variance in satisfaction with participation was explained by fatigue together with feelings of feelings of depression.

## DISCUSSION

Patients with early onset SMA (type 1,2,3a) experienced more participation restrictions than patients with later onset SMA, but reported similar levels of frequency of participation and resulting satisfaction. Motor skills were independently associated with the frequency of and

restrictions in participation. In addition, subjective complaints, namely fatigue, pain and particularly feelings of depression, were associated with participation scores in bivariate analyses. Coping styles were not associated with participation scores. In the multiple regression analyses, having feelings of depression proved to be the only subjective complaint independently related to participation (frequency and satisfaction).

This study identifies the participation activities that pose most problems for adult patients with SMA. There are a few studies on specific domains of participation that enrolled patients with a broader range of neuromuscular diseases. Employment rates of 40-57 % were, for example, found in a mixed sample of patients with neuromuscular disorders.[12,13,14] This percentage is not only similar to our findings, but also comparable to that of patients with spinal cord injury.[28] Our data, therefore, seem to be in line with previous reports on participation in work activities of patients with moderate to severe motor impairments. Satisfaction scores of patients with SMA were also similar to those previously reported by patients with spinal cord injury and patients after stroke.[28,29] It reflects the fact that patients in different situations are able to reorganize their social activities on average, in a satisfactory way, regardless of their restrictions and type of condition.

Age at onset of SMA may also be relevant for satisfaction with participation, since patients with SMA type 1c-3a experienced more restrictions but similar levels of satisfaction as patients with a later onset. A possible explanation for this finding is that patients with early onset SMA might have adapted more successfully to living with severe physical limitations from a very early age onwards.[43] Patients with late onset SMA initially have normal motor development. Relevant physical limitations due to disease progression occur in adulthood. Continuous adaptation attempts might lead to enduring distress as these patients have to redefine their goals and concepts about their daily functioning.

Although the incidence of feelings of depression among patients with SMA is not higher than in those with other neuromuscular diseases,[18,20,21] the presence of feelings of depression was inversely associated with both frequency of participation activities and satisfaction with participation. This suggests that it is important to monitor whether feelings of depression are present in adult patients with SMA, and if so, to consider psychological interventions in order to reduce these feelings that might be the result of impaired emotional adaptation to disease progression.[43,44]

Coping style was not associated with any aspect of participation. To the best of our knowledge, there are no studies on the relationship between coping style and social participation in patients with neuromuscular disorders. Studies have, however, been carried out on MS patients and spinal cord injury patients, and these also failed to find a relationship between coping styles and participation.[45,46] The fact that healthy individuals exhibit higher levels of coping variability than patients with chronic disease may play a role in this.[47] We cannot, however, exclude the possibility that coping styles are relevant for participation (satisfaction) in a subgroup of patients, in particular among patients with SMA type 3-4 who may face challenges of adaptation in later life.

As in other studies, pain was reported frequently, in particular by late onset patients.[18,48] Causes of pain are multiple, and include spinal deformities, muscle cramps or neurogenic pain. Univariable analysis showed that pain is associated with the frequency of participation activities in patients with SMA, but when feelings of depression was entered into the model, pain did not add significantly to the model (see table 6).

The limited sample size is an important limitation of this study. It allowed the (pre)selection of a limited number of possible correlates. We made this preselection based on the existing literature and clinical experience. In our study we included adult patients with SMA over the whole spectrum (type 1c-4) and assessed participation in more detail than before. This gave

us the opportunity to assess the relevance of already known variables in a broader group of patients with SMA, thus expanding the clinical relevance of our study. Follow-up studies should aim at a larger sample size to address the importance of other factors including endurance and stamina for motor activities, social support, upper extremity function and personal factors such as self-efficacy or illness perceptions, in particular since the largest portion of the variance in participation remains unexplained. Larger sample sizes would also allow more detailed subgroup analysis, for example, of subgroups with early and later onset. Although almost 80% of invited patients participated in this population-based study, we cannot fully exclude the possibility of inclusion bias, i.e., the selection of patients in a relatively good condition or of patients who experienced increasing problems (e.g. patients with SMA type 3b), which may have influenced the results of this study.

In conclusion, this study showed that although less restricted, patients with late onset SMA do not feel greater satisfaction with their participation in daily life than patients with early onset SMA. Compared to other diagnoses (e.g. spinal cord injury), SMA patients appear to be as satisfied with their participation in daily activities. Late onset patients reported more fatigue and experienced more pain than patients with early onset SMA. Motor skills, fatigue and feelings of depression in particularly are correlates of participation in daily life. Although these findings do not fully explain variation in participation, addressing these problems may be helpful in optimizing and personalising rehabilitation care for adult patients with SMA.

**Abbreviations:**

| | |
|---|---|
| SMA | spinal muscular atrophy |
| SMN gene | survival motor neuron |
| USER-P | Utrecht Scale of Evaluation Rehabilitation-Participation |
| HFMSE | Expanded Hammersmith Functional Motor Scale |
| SF-36 | Short Form 36-item Health Survey |
| HADS | Hospital Anxiety and depression Scale |
| CISS-21 | Coping Inventory for Stressful Situations (Short version) |

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Table 1. Demographics in the total population and in 2 subgroups, early versus late onset SMA

| | **Total** | **Type 1** | **Type 2** | **Type 3a** | **Type 3b** | **Type 4** | **Early onset** | **Late onset** |
|---|---|---|---|---|---|---|---|---|
| N | 62 | 4 | 21 | 13 | 20 | 4 | 38 | 24 |
| Sex, female | 55% | 75% | 62% | 62% | 45% | 60% | 63% | 50% |
| Age in years | 43.0 (20-70) | 40.5 (24-51) | 31.0 (20-68) | 51.0 (20-66) | 48.0 (20-70) | 53.0 (43-69) | 37.0 (20-68) | 48.0 (20-70) |
| Partner | 61% | 25% | 38% | 69% | 80% | 100% | 47%* | 83% |
| Paid job | 39% | 25% | 29% | 31% | 55% | 50% | 30% | 54% |
| Education | | | | | | | | |
| - No /Secondary school | 66% | 50% | 72% | 54% | 65% | 100% | 64% | 71% |
| - Higher education | 34% | 50% | 29% | 46% | 35% | 0 | 37% | 29% |
| Living independent | 68% | 75% | 38% | 85% | 80% | 100% | 58% | 83% |

Note: values are median (range). *Results differ significantly between groups (SMA types or early versus late onset) with $p<0.05$. NA, not applicable. Education, highest grade level completed; Partner: living separate or together = yes; Living independent: independent with/without self-coordinated care = yes; Paid employment: frequency scale USER-P, Yes/No;

Table 2. Disease severity and psychological factors in the total population and in 2 subgroups, early versus late onset SMA

| | **Total** | **Type 1** | **Type 2** | **Type 3a** | **Type 3b** | **Type 4** | **Early onset** | **Late onset** |
|---|---|---|---|---|---|---|---|---|
| N | 62 | 4 | 21 | 13 | 20 | 4 | 38 | 24 |
| **Disease severity** | | | | | | | | |
| Motor skills (HFMSE) | 3.5 (0-66) | 0 | 2.0 (0-20) | 4.0 (0-35) | 29.0 (0-66) | 48.0 (43-53) | 2.0* (0-35) | 32.5 (0-66) n=22 |
| Age at diagnosis | 1.5 (0-44) | 0.42 (0.33-0.5) | 0.75 (0-2) | 1.5 (0.5-3) | 6.5 (3.5-25) | 40.3 (31-44) | 0.95 (0-3)* | 9.5 (4-44) |
| Loss ambulation | 18.5 (2-59) | NA | NA | 17.6 (4-46) | 35.1 (9-59) | NA | 13.9* (1-45) | 34.5 (9-59 |
| **Subjective complaints** | | | | | | | | |
| Pain (SF-36) | 84.0 (0-100) | 73.0 (61-84) | 84.0 (20-100) | 84.0 (31-100) | 57.0 (0-100) | 73.0 (41-100) | 84.0* (20-100) | 62.0 (0-100) |
| Vitality (SF-36) | 62.5 (5-100) | 70.0 (45-75) | 70.0 (30-100) | 80.0 (5-100) | 55.0 (15-70) | 57.5 (45-95) | 75.0* (5-100) | 55.0 (15-95) |

| | | | | | | | | |
|---|---|---|---|---|---|---|---|---|
| HADS-A | 3.5 (0-21) | 4.5 (3-9) | 4.0 (0-10) | 3.0 (0-9) | 3.0 (0-21) | 2.5 (2-3) | 4.0 (0-10) | 3.0 (0-21) |
| HADS-D | 2.0 (0-15) | 2.0 (1-4) | 1.0 (0-9) | 2.0 (0-15) | 3.5 (10-12) | 2.0 (1-5) | 1.5 (0-15)* | 3.0 (1-12) |
| **Psychological factor** | | | | | | | | |
| CISS: | | | | | | | | |
| - Task-oriented | 26.6 (12-33) | 25.5 (19-32) | 26.4 (12-33) | 25.7 (12-33) | 27.8 (20-33) | 24.7 (19-28) | 27.0 (12-33) | 28.0 (19-33) |
| - Emotional | 14.6 (7-32) | 12.5 (8-15) | 15.3 (7-25) | 12.5 (7-19) | 15.7 (7-32) | 14.7 (11-18) | 14.1 (7-25) | 15.5 (7-32) |
| - Avoidance | 19.3 (7-31) | 22.0 (16-27) | 18.9 (12-28) | 19.1 (7-31) | 18.7 (9-27) | 21.3 (11-28) | 19.5 (7-31) | 19.1 (9-28) |

Note: values are median (range). *Results differ significantly between groups (SMA types or early versus late onset) with $p<0.05$. NA, not applicable.

Loss of ambulation: stop walking with aid, in years.

Abbreviations: SMA, Spinal muscular Atrophy; HFMSE, Expanded Hammersmith Functional Motor Scale; MRC, Medical Research Council; SF-36, Short Form 36–item Health Survey; HADS, Hospital Anxiety and Depression Scale; HADS-A, Hospital Anxiety and Depression Scale; anxiety subscale; HADS-D, Hospital Anxiety and Depression Scale; depression subscale; CISS-21, Short Form of the Coping Inventory for Stressful Situations

Table 3. USER-P scores for the total population, and early versus late onset SMA

| | | **SMA type** | | | | | **Onset** | |
|---|---|---|---|---|---|---|---|---|
| | Total | Type 1 | Type 2 | Type 3a | Type 3b | Type 4 | Early | Late |
| USER-P | N=62 | n=4 | n=21 | n=13 | n=20 | n=4 | n=38 | n=24 |
| Frequencies total | 34.5 (7-66) | 32.1(21-43) | 36.7 (17-49) | 30.9 (7-50) | 33.5 (18-48) | 42.2 (23-66) | 35.4 (7-50) | 35.4 (18-66) |
| - Frequency: work/education | 20.0 (0-45) | 17.5 (10-40) | 25.0 (0-40) | 10.0 (0-30) | 22.5 (0-45) | 15.0 (5-35) | 20.0 (0-40) | 20.0 (0-45) |
| - Frequency: leisure activities | 48.6 (14-74) | 42.9 (31-54) | 54.3 (31-71) | 48.6 (14-74) | 44.3 (26-74) | 55.7 (40-65) | 50.0 (14-74) | 47.1 (25-74) |
| Restrictions | 61.9 (18-100) | 40.6 (33-59) | 58.2 (36-93) | 61.1 (18-89) | 67.1 (30-97) | 79.4 (59-100) | 57.6 (18-93)* | 73.3 (30-100) |
| Satisfaction | 72.5 (32-100) | 71.9 (47-86) | 72.5 (50-100) | 77.8 (32-100) | 71.0 (33-89) | 74.3 (58-98) | 75.0 (32-100) | 71.0 (33-98) |

Note: values are median (range).

*Results differ significant between groups (SMA types or early versus late onset) with $p<0.05$

SMA type, SMA types 1-4; Onset, early versus late onset SMA.
Abbreviations: SMA, Spinal Muscular atrophy; USER-P, Utrecht Scale for Evaluation Rehabilitation-Participation.
Frequency, work/education: hours/ week; Frequency, leisure activities: times/month.

Table 4. USER-P, Frequency scale: hours per week (work/education) and times per month (leisure activities) spent per item, for the total population, N = 62

| **Domains** | | **Hours/week** | |
|---|---|---|---|
| | Not at all | 1-24 hrs | ≥25 hrs |
| Paid work | 61% | 18% | 21% |
| Unpaid work | 44% | 42% | 15% |
| Education | 71% | 18% | 12% |
| Household | 34% | 60% | 7% |
| | | **Times/month** | |
| | Not at all | 1-10 times | >10 times |
| Sports and physical exercise | 44% | 42% | 15% |
| Going out | 10% | 87% | 2% |
| Daytrips | 11% | 81% | 8% |
| Leisure activities at home | 5% | 39% | 57% |

| | | | |
|---|---|---|---|
| Visiting family or friends | 3% | 86% | 11% |
| Receiving visitors | 3% | 87% | 10% |
| Contact phone, computer | 0 | 16% | 84% |

Note: Education: only activities in the course of work, or in order to get work.
Abbreviations: USER-P, Utrecht Scale for Evaluation Rehabilitation-Participation

Table 5. Percentage of patients with perceived restrictions and dissatisfaction, for the total population, N=62

| **Restrictions** | **%** | **Dissatisfaction** | **%** |
|---|---|---|---|
| **Persisting problems** | | | |
| Work/education | 67 % (n=52) | Work/education | 15 % (n=39) |
| Household chores | 97 % (n=59) | Household chores | 43 % (n=56) |
| Mobility outdoors | 82% | Mobility outdoors | 31 % (n=61) |
| Physical exercise | 95 % (n=57) | Physical exercise | 58 % (n=50) |
| Going out | 71% | Going out | 31 % (n=61) |
| Outdoor activities | 79% | Outdoor activities | 31 % (n=61) |
| Leisure indoors | 41 % (n=61) | Leisure indoors | 12 % (n=61) |
| Relationship partner | 33 % (n=40) | Partner relationship | 7 % (n=39) |
| Visits to family/friends | 66% | Family relationships | 10% |
| Visits from family/friends | 20 % (n=61) | Friends and acquaintances | 8% |
| Telephone/computer contact | 16 % | | |

(n..): n per item NB: Not all items were scored by all patients.

Table 6. Bivariate and multivariable linear regression analyses for USER-P frequency, restrictions and satisfaction. Stepwise regression.

| | **Frequency of participation** | | | **Participation restrictions** | | | **Satisfaction with participation** | | |
|---|---|---|---|---|---|---|---|---|---|
| | Bivariable | Multivariable | | Bivariable | Multivariable | | Bivariable | Multivariable | |
| | | Step 1 | Step 2 | | Step 1 | Step 2 | | Step 1 | Step 2 |
| **Characteristics** | ρ | β | β | ρ | β | β | ρ | β | β |
| Sex, female | 0.08 | N.E. | N.E. | 0.05 | N.E. | N.E. | -0.03 | N.E. | N.E. |
| Age in years | -0.28* | -0.24 | -0.05 | -0.10 | N.E. | N.E. | -0.01 | N.E. | N.E. |
| Partner | 0.03 | N.E. | N.E. | 0.11 | N.E. | N.E. | 0.06 | N.E. | N.E. |
| Education, High | 0.05 | N.E. | N.E. | 0.06 | N.E. | N.E. | 0.32* | -0.37 | -0.20 |
| Living independent | -0.13 | N.E. | N.E. | -0.01 | N.E. | N.E. | -0.13 | N.E. | N.E. |
| Motor skills (HFMSE) | 0.26* | 0.27* | 0.32* | 0.59* | 0.45* | 0.47* | -0.04 | N.E. | N.E. |
| Pain (SF-36) | 0.28* | N.E. | 0.06 | 0.04 | N.E. | N.E. | 0.21 | N.E. | N.E. |

| | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|
| Fatigue (SF-36) | 0.19 | N.E. | N.E. | -0.01 | N.E. | N.E. | 0.42* | N.E. | 0.30* |
| HADS-A | 0.01 | N.E. | N.E. | -0.05 | N.E. | N.E. | -0.29 | N.E. | N.E. |
| HADS-D | -0.41* | N.E. | -0.44* | -0.24* | N.E. | -0.23 | -0.49* | N.E. | -0.41* |
| CISS | | | | | | | | | |
| Task-oriented | -0.06 | N.E. | N.E. | -0.14 | N.E. | N.E. | -0.03 | N.E. | N.E. |
| Emotional | 0.21 | N.E. | N.E. | -0.11 | N.E. | N.E. | -0.00 | N.E. | N.E. |
| Avoidance | -0.09 | N.E. | N.E. | -0.05 | N.E. | N.E. | 0.02 | N.E. | N.E. |
| $\Delta R^2$ | | 0.14* | 0.19* | | 0.21* | 0.05 | | 0.14* | 0.36* |
| **$R^2$** | | **0.33** | | | **0.26** | | | **0.50** | |

Note: $\Delta R^2$, change in $R^2$ between two equations; $R^2$**,** explained variance.

*, all included variables significant correlation with outcome measure $p<0.1$.

Abbreviations: USER-P, Utrecht Scale for Evaluation Rehabilitation-Participation; HFMSE, Expanded Hammersmith Functional Motor Scale; SF-36, Short Form 36–item Health Survey; HADS-A, Hospital Anxiety and Depression Scale; anxiety subscale; HADS-D, Hospital Anxiety and Depression Scale; depression subscale; CISS-21, Short Form of the Coping Inventory for Stressful Situations.